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Orbito-nasal cyst in a young European short-haired cat

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Running title: Orbito-nasal cyst in a young European short-haired cat

ABSTRACT

Purpose To describe a case of an orbito-nasal cyst in a cat.

Procedure An eighteen months old male European short-haired cat was presented to the Ophthalmology service of the Vetsuisse Faculty, University of Zurich for a subcutaneous swelling in the medial canthal region of the right eye (OD). Ophthalmologic, ultrasound and CT examinations, and fine needle aspiration were performed. After lesion excision the removed tissue was submitted for histopathology. CT examination was repeated 5 months after removal of the cyst.

Results Ophthalmologic examination revealed a large fluctuant swelling inferonasal to OD. Despite patent lacrimal puncta, only the first few mm of the lacrimal canaliculi could be cannulated. A normal globe with moderate enophthalmos was present. Ultrasound examination showed a well-defined lobulated cyst-like structure in the right orbit, inferonasal and anterior to the eye. CT examination revealed extension of this lesion through the medial orbital wall into the right nasal cavity. Fine needle aspiration confirmed the cystic nature of the lesion. An orbito-nasal cyst was diagnosed. The orbital part of the cyst was dissected from the surrounding tissue and excised from the periosteum in the medial orbital wall defect. Part of the maxillary bone was removed to allow removal of the cyst from the nasal cavity. Histologically, the cyst wall consisted of a single to multi-layered, mostly cuboidal epithelium and surrounding connective tissue. Follow-up revealed a good functional result and no recurrence 7 months after cyst removal.

Conclusions Similar orbito-nasal cystic structures were reported in dogs, but not in cats.

Key Words: orbito-nasal cyst, cat, CT, NL duct, excision, histopathology

INTRODUCTION

Cystic lesions in the periorbital area are rare in domestic animals.(1) In dogs, reported cases include dacryops,(1, 2, 3, 4) congenital medial canthal cyst,(5) canaliculops,(6) cystic dilatation of the nasolacrimal (NL) duct,(7, 8) maxillary bone epithelial cyst,(9, 10) suspected epithelial cyst,(11) periorbital epidermoid cyst,(12) cholesterol granuloma,(13) zygomatic salivary mucocele and cysts of the frontal and nasal sinuses.(1)

A dacryops is a cyst originating from lacrimal glandular ductal tissue, which can be confirmed histologically.(2, 3) Specifically, myoepithelial cells, which are present in tissues around lacrimal gland acini and ductules, can be detected via immunohistochemical staining for smooth-muscle actin. This was recently described in a report of 3 cases of dacryops in young Labrador retrievers by Ota *et al.*(2)

This case report describes the clinical presentation, diagnosis, surgical treatment and post-operative follow-up of an orbito-nasal cyst in a cat.

CASE REPORT

Case history

An 18 months old, intact male, domestic shorthaired cat was presented to the Ophthalmology service of the Vetsuisse Faculty, University of Zurich, Switzerland, for the evaluation of a subcutaneous swelling in the medial canthal area of the right eye. The cat belonged to the Institute for Animal Nutrition at the Vetsuisse Faculty of Zurich, and was engaged in a nutritional study. The exact duration of this swelling was unknown, but mild swelling had already been noticed when the cat was a kitten, and had started to enlarge over the last several months. According to the caretaker, the swelling was not associated with any discomfort but the eye had frequently been tearing since the cat was a few months old. There was no known history of trauma.

Previous problems associated with this eye include conjunctivitis, keratitis and lower eyelid entropion. A PCR for detection of FHV-1 DNA from a cytobrush sample taken from the lower conjunctival fornix had been performed previously at a time when the cat also demonstrated signs of rhinitis. Although the result was negative, FHV-1 was suspected as the etiologic agent for the conjunctival and corneal inflammation. Also, the lower eyelid entropion was suspected to be the result of longstanding blepharospasm, due to painful herpetic ocular surface disease. Since entropion did not resolve after temporary tarsorrhaphy, permanent correction with a Hotz-Celsus procedure was performed, which restored a normal lower eyelid position.

Clinical findings

The results of a neuro-ophthalmological examination, including menace response, dazzle, pupillary and palpebral reflexes were normal. A well-circumscribed, oval fluctuant subcutaneous swelling was present inferonasal to the right eye. (Fig 1) The swelling was non-movable, was not warm or painful, but did appear tender on firm palpation. There was some dried mucoid discharge on the eyelids, especially in the medial canthus, but the skin overlying the swelling was unremarkable. No obvious epiphora was present. The eyelid fissure was narrower compared to that of the other eye. Motility of the lower eyelid was impaired due to the swelling, but the eyelid fissure could still be closed completely. The right globe was moderately enophthalmic, and obvious resistance was felt on retropulsion in a ventral and medial direction. Retropulsion in other directions was unremarkable. No pain was elicited with retropulsion or opening of the mouth.

Eyelid margin position was appropriate, but there was some fibrosis of the palpebral conjunctiva in the medial canthus, including the area of the lower lacrimal punctum. With the lower eyelid in an everted position, the swelling could also be appreciated beneath the nasal palpebral conjunctiva.

Passage of fluorescein dye through the nasolacrimal duct (Jones test I) was absent OD. The lacrimal puncta OD could be cannulated, but the cannula could only be advanced for one to two mm into the lacrimal canaliculi. The NL system could therefore not be irrigated.

The palpebral conjunctiva was mildly hyperemic. Mild fibrosis and fine ghost vessels were present in the inferotemporal cornea, but no retainment of fluorescein stain was observed.

The rest of the ophthalmic examination of the right eye, including slit-lamp biomicroscopy, indirect ophthalmoscopy and applanation tonometry, did not reveal any abnormalities. The examination of the left eye was unremarkable. Beside mild mucopurulent discharge around the right nostril, the physical exam was unremarkable.

Preoperative imaging

On ultrasound examination of the right orbital area, a lobulated structure with a well-defined contour and anechoic content was observed in contact with the inferonasal aspect of the right globe. The morphology and content of this structure was consistent with a cyst. The right globe was unremarkable. (Fig 2)

Upon aspiration of the cyst, a slightly opaque, brownish fluid was obtained. The cyst fluid had a specific gravity of 1.006, protein content of < 1 g/l and cell count of 13/μl. Cytological evaluation of the cyst fluid revealed a moderate number of erythrocytes and few neutrophils and small lymphocytes.

Based on the results of the clinical examination, orbital ultrasound and analysis of aspirated fluid, an orbital cyst was diagnosed.

In order to evaluate the extension and possible associated bone involvement, a CT scan of the head was performed with the animal under anesthesia. The CT study was done using a multi-row unit[#], with a beam collimator of 40 data channels with a detector-row width of 0.6 millimeters (40 x 0.6 mm), a tube potential of 120 kilovolts (peak kilovoltage) and a tube

[#] Somatom Sensation Open , Siemens AG, Medical Solutions, Erlangen, Germany

current of 200 milliamperes. Images were reconstructed with a section thickness of 0.75 mm, using both a soft tissue and a bone window. (Fig 3) A post contrast scan was obtained 30 seconds after the intravenous injection of an iodinated high osmolar contrast medium ** with a dose of 700 mgI/kg and a concentration of 350 mgI/ml. The CT images revealed a severe enlargement of the osseous component of the lacrimal canal. Additionally, an ovoid structure with soft tissue density and well-defined contours extended from the ventromedial aspect of the eye, through the widened lacrimal canal and into the nasal cavity. This structure displaced the right eye caudoventrally and the ventral nasal conchal bone axially. Three-dimensional bone reconstructions are shown in Fig 4. For 3-dimensional reconstructions, the pixel values of the cyst have been changed to demonstrate the location and extension of the cyst. Attempts to identify and cannulate the nasal ostium of the NL duct were unsuccessful. Because the lacrimal canaliculi could not be cannulated either, dacryocystorhinography could not be performed.

Since the cyst was expanding significantly, progressively dislocating the globe and obstructing the visual axis, and the cyst refilled within days after fluid aspiration, a surgery to remove the cyst was recommended and scheduled.

Surgery

The cat was premedicated with ketamine hydrochloride (8 mg/kg of body weight (BW); Narketan[®], Vétoquinol) in combination with midazolam (0.1 mg/kg BW; Dormicum[®], Roche) administered intramuscularly. Induction was then performed with propofol (5 mg/kg BW IV; Diprivan[®], AstraZeneca). After intubation, the cat was maintained on isoflurane (AttaneTM Isoflurane, Provet) and oxygen. Amoxicillin (20 mg/kg BW; Clamoxyl[®], Pfizer) was administered intravenously. A presurgical clip was performed on the right side of the head and the surgical field was aseptically prepared. The cat was positioned in left lateral recumbency.

** Telebrix[®] , 350 mgI/ml, Guerbert, Zurich, Switzerland

147 A 3 cm skin incision was performed 1.5 cm below and parallel to the lower eyelid margin.
148 The orbital part of the cyst was bluntly dissected from the surrounding tissues. (Fig 5) During
149 dissection a fibrous strand of tissue, running from the dorsal cyst wall to the palpebral
150 conjunctiva close to the lower nasal eyelid margin, was identified. A small perforation of the
151 cyst wall, resulting in collapse of the cyst, occurred when the dissection approached the
152 periosteum of the maxillary bone. To facilitate the rest of the dissection, the cyst was filled
153 with 1.2% hyaluronic acid (Biovisc 1.2%[®], Acrivet). The cyst was then excised at the level of
154 the periosteum of the inferonasal orbital rim, revealing an oval defect (maximal diameter: 12
155 mm) in the lacrimal and maxillary bone. (Fig 6) The extension of the cyst into the nasal
156 cavity could be identified through this defect. Since the cyst in the nasal cavity expanded well
157 beyond the margin of the bone defect, this opening needed to be enlarged in order to remove
158 the cyst. This was accomplished in two steps. First, the compact bone was removed with use
159 of a pneumatic bone drill. Then, the rest of the bone was removed with angled Cicherelli and
160 Kerrison rongeurs. Cyst tissue was removed from the nasal cavity, enabling direct
161 visualization of nasal turbinates. The cyst was placed in 10% neutral buffered formalin and
162 submitted for histologic examination. Moderate bleeding occurring during the procedure was
163 controlled with oxidized cellulose (Tabotamp[®], Johnson & Johnson). The wound was closed
164 with single interrupted sutures in three layers: the fascia edges were apposed using 3-0
165 polydioxanone (PDS[®], Ethicon) suture in an interrupted cruciate pattern. Subcutis (4-0
166 poliglecaprone, Monocryl[®], Ethicon) and skin (4-0 polyamide, Supramid[®], Braun) were
167 closed routinely with simple interrupted sutures. No drain was inserted.

168 At the conclusion of surgery, meloxicam (0.1 mg/kg BW; Metacam[®], Boehringer Ingelheim)
169 and buprenorphine (0.015 mg/kg BW; Temgesic[®], Schering-Plough) were administered
170 intravenously for postoperative analgesia. Recovery from anesthesia was uneventful. The cat
171 was released from the hospital the same day, with the following therapy: amoxicilin /
172 clavulanic acid (Clavubactin[®], Dr. E. Graeub) 20 mg/kg BW BID PO for 14 days, meloxicam

oral suspension for cats (Metacam[®], Boehringer Ingelheim) 0.1 mg/kg BW once daily for the first day after the surgery, followed by 0.05 mg/kg BW once daily for 5 more days, 0.3% tobramycin eyedrops (Tobrex[®], Alcon) and lubricating gel with Vitamin A (Oculotect[®], Novartis) QID OD for 14 days. An Elizabethan collar was recommended for 14 days until removal of the external sutures.

Histology

Examination of sections stained with haematoxylin and eosin (HE) demonstrated a cyst wall consisting of a single or double and rarely multiple layers of mostly cuboidal epithelial cells forming a stratified epithelium surrounded by a highly cellular connective tissue capsule. No goblet cells or cilia could be detected. Multiple deposits of well-mineralized metaplastic bone and lymphocytic aggregates were present in the surrounding connective tissue. (Fig 7) Immunohistological staining for smooth-muscle actin was performed and was found negative (data not shown). The histological diagnosis was a non neoplastic cyst.

Follow up

The wound had healed nicely on recheck three weeks after surgery. (Fig 8) Mild enophthalmus and nictitating membrane protrusion were present and the palpebral fissure was narrower compared to that of the other eye. The palpebral reflexes were normal, although the motility of the lower lid was still mildly impaired. No difference in retropulsion was present between both eyes. Also, no pain was elicited with palpation of the eyelids and retropulsion of the globe. No epiphora was present, but some dried, mucoid discharge was present around the right nostril. No stridor was detected. No other changes from previous examinations were observed in either eye. No further therapy was prescribed.

On recheck 5 months after the cyst removal, the right palpebral fissure was mildly narrowed and mild epiphora from the right eye was present. There was mild entropion without trichiasis

of the lower eyelid margin on the operated side, and no discomfort was noticed by the examiners, nor reported by the handler. The rest of the ocular exam was unremarkable in both eyes.

Postoperative imaging

In the same follow-up examination, 5 months after the surgery, the cat was anaesthetized as previously described, and the CT examination was repeated. The previously identified cyst-like structure could not be identified. (Fig 9) There was a persistent mild caudal and ventral displacement of the right eye. The widened osseous component of the lacrimal canal was unchanged compared to the pre-operative study. The right ventral conchal bone presented a normal position and the left ventral nasal concha was filled by a soft tissue dense material. This finding was interpreted as rhinitis. Additionally, a defect on the tip of the root of the upper right canine tooth was identified. This lesion was likely the consequence of iatrogenic trauma during maxillary bone removal.

The following options were given by the Dentistry service: recheck with dental radiographs in 4 months, apical preparation with retrograde filling of the root canal or extraction of the tooth. The prognosis of the retrograde filling procedure would be guarded at best due to various reasons. The handler has opted for observation and regular rechecks, and would decide for an extraction of the tooth in case of any problems.

On the last recheck, 7 months after surgery, no changes from the previous exam were noticed. No pain or ocular discomfort was reported by the caretaker, but constant mild tearing from this eye was present. (Fig 10)

DISCUSSION

Periorbital cysts are rare in domestic animals,(1) and may originate from several glandular or ductular structures, including the orbital lacrimal gland, lacrimal gland of the nictitating

221 membrane, zygomatic salivary gland, lacrimal canaliculi, sac and/or NL duct, conjunctival
222 goblet cells, frontal and nasal sinus mucosa, and transposed parotid duct.(1, 6) The etiology of
223 these cysts is various, and includes congenital malformation, trauma, inflammation and
224 neoplasia.(1) In the report by Martin *et al.*, the anatomic location of the cyst and any
225 associated lesions was stated to be the most consistent clue to its origin.(1)

226 This report describes the case of an orbito-nasal cyst in a young cat. The origin of the cyst, as
227 well as its etiology, remains unknown. Due to the anatomic location, it is very likely that the
228 cyst was closely associated with, or originated from, the lower lacrimal canaliculus or
229 nasolacrimal duct.

230 Although we could not find any reports of orbito-nasal cysts in cats, there are reports
231 describing similar conditions in dogs.

232 A case of a nasal and orbital lacrimal cyst, accompanied by occlusion and dilatation of the NL
233 duct, was described by Grahn *et al.* in a six months old basset hound.(3) White *et al.*
234 described a case of a cystic obstruction of the nasolacrimal duct in a one year old
235 dachshund.(7) Ito *et al.* reported a cyst, suspected to be epithelial, that occupied a defect in
236 the maxillary, lacrimal and frontal bones and invaded the nasal cavity in a 4-year old
237 miniature schnauzer.(11)

238 Although the clinical presentation in these cases was similar in that cystic structures affecting
239 the orbit and the nasal cavity with a defect in the orbital wall were present, there were some
240 differences regarding the tissue of origin and possible etiology of the cyst. In the report by
241 Grahn *et al.*, the seromucinous glandular tissue identified in the cyst wall most likely
242 originated from the third eyelid gland, and a congenital lesion was suspected. The NL duct
243 was occluded by the cyst, and dilatation of the duct might have been related to continuous tear
244 flow.(3) In the case described by White *et al.*, the cyst consisted of the distended NL duct due

to distal obstruction by a chronic inflammatory process. The specific cause for the inflammation was not identified.(7) In the report by Ito *et al.*, the etiology was unknown, and although no abnormalities of the NL duct were reported, the duct was still thought to be the most likely structure of origin.(11)

Dacryocystorhinography is a radiographic contrast examination procedure used to delineate the nasolacrimal drainage system and evaluate its patency.(14-16)

Since it was not possible to cannulate the lacrimal canaliculi or the nasal ostium of the NL duct, dacryocystorhinography could not be performed in this case. Therefore, eventual communication of the lumen of the cyst with the lumen of the NL system could not be ascertained. But even if dacryocystorhinography could have been performed, and the dacryocystorhinogram would not have identified a communication, a connection of the cyst wall with the NL duct could not have been ruled out. During surgery, a fibrous strand of tissue was found connecting the dorsal cyst wall and the palpebral conjunctiva close to the lower medial eyelid margin corresponding to the site of the lower lacrimal punctum. Although we cannot support this histopathologically, we think that this structure was most likely a fibrosed lower lacrimal canaliculus, and that the cyst was actually connected with the NL system.

Based on negative immunohistochemical staining for α -smooth actin, we conclude that the cyst did not originate from lacrimal gland tissue or ductules,(2) which supports our assumption of a canalicular origin for the cyst.

To the best of our knowledge, there is no information in the literature regarding the histology of the NL duct in cats. In the dog, a bistratified columnar anatomy has been reported for the epithelium of the lacrimal sac and the proximal and middle portion of the nasolacrimal duct.(17) Identification of a single to multilayered, mostly cuboidal epithelium lining the cyst in our case, does not exclude the possibility that the cyst did originate from the NL system.

269 Also, this finding makes it very unlikely that this was an epidermoid or dermoid cyst, both of
270 which are characterized by a stratified squamous epithelium, or a cholesterol granuloma
271 which lacks the epithelial layer.(9, 12, 13)

272 The defect in the orbital wall could be explained by bone atrophy due to pressure exerted by
273 the enlarging cyst as was described previously.(7, 9)

274 The nasal conchae, which were displaced by the cyst on preoperative CT, were found to be
275 reorganizing on the postoperative CT.

276 We hypothesize that the previously diagnosed conjunctivitis, keratitis and entropion were
277 caused by FHV-1. The fibrotic changes of the palpebral conjunctiva and lacrimal canaliculi
278 were interpreted as symblepharon, which is a well known sequel of FHV-1 induced ulcerative
279 conjunctivitis.(18)

280 Despite the negative result of a PCR assay for the detection of FHV-1 DNA, FHV-1 should
281 not necessarily be ruled out as the etiologic agent for the above mentioned changes.(19) Many
282 cats which are likely to have FHV-1 associated disease display negative PCR results.(20)

283 It is possible that viral DNA was not present in the tested sample or was not present in
284 detectable amounts.(19, 21) Alternatively, the cat might not have been shedding virus at the
285 time of sampling.(19)

286 It is impossible to say whether the nasal ostium of the NL duct was actually absent or we were
287 just unable to localize it.

288 Removal of the cyst was advised and performed because the increasing size of the cyst was
289 causing globe displacement and obscuration of the visual axis. Also, the cyst refilled quickly
290 after aspiration of the fluid and further enlargement of the cyst was therefore expected.

291 Possible treatment alternatives have been reported in the literature. The intralesional

application of tetracycline has been mentioned as a fibrosing agent for the treatment of periocular cysts, but at the same time was not recommended by the author because of possible conjunctival necrosis and severe periocular inflammation.(22) Sclerotherapy with ethanalamine oleate was recently described for the management of an orbito-palpebral cyst associated with congenital microphthalmos of a non-visual eye.(23) The use of polidocanol, another sclerosing agent, has been reported anecdotally as therapy for dacryops in dogs, but no reports have been published to this date. Due to the size and extension of the cyst, the possible side effects of a treatment with sclerosing agents and the lack of publications supporting the use of such a therapy, chemical ablation of the cyst was not attempted in this case and surgical excision was performed instead.

No recurrence of the cyst was noticed on repeat CT examination 5 months after surgery. Mild enophthalmos, a narrowed palpebral fissure, epiphora, mild inversion of the lower eyelid margin and nictitating membrane protrusion were noticed on rechecks 5 and 7 months after cyst removal. The presence of enophthalmos, nictitating membrane protrusion and an abnormal eyelid position might be attributed to the osseous defect in the medial orbital wall and/or atrophy of the orbital fat due to the pressure exerted by the enlarging cyst. No discrepancy in the size of the right and left orbit or globe was noticed on the CT images. Protrusion of the nictitating membrane, both before and after surgery, could also be attributed to symblepharon, although no obvious restriction of nictitating membrane motility was noticed during or after cyst removal. Epiphora was thought to be caused by a combination of absent tear drainage through the nasolacrimal system and presence of mild tipping in of the lower eyelid margin. Since no trichiasis was present and no discomfort was noticed at recheck examinations nor reported by the handler, correction of this mild entropion was judged to be unnecessary. The injury of the root of the upper canine tooth that occurred during the removal of bone was accidental, and was not noticed intraoperatively. Considering the anatomical location of the tooth, it is possible that trauma to the tooth root occurred unnoticed, and this

emphasizes the importance of extreme caution during removal of bone in such proximity to the teeth.

Since this is an intact male cat, which at least occasionally had contact with other cats, ocular trauma cannot be ruled out completely as contributing factor in the cyst formation. Also, considering the young age of the cat, a congenital abnormality is possible.

In conclusion, the presentation, diagnosis, removal and long-term follow-up of an orbito-nasal cyst are described. The origin and etiology of the cyst remain unknown. Removal of the cyst resulted in a good functional outcome and lack of recurrence 7 months after surgery.

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